Rupture of a Large Vertebral Artery Aneurysm Following Proximal Occlusion

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Summary

Proximal occlusion of the vertebral artery is regarded as a safe and effective method of treating aneurysms of the vertebral artery or the vertebrobasilar junction unsuitable for treatment by neck clipping. Complications known to develop after this procedure include ischemic lesions of the perforators and other areas. There are only a limited number of reports on early rupture of aneurysm following proximal occlusion of the vertebral artery for the treatment of unruptured aneurysm. We recently encountered a case of large aneurysm of the vertebral artery identified after detection of brainstem compression. This patient underwent proximal occlusion of the vertebral artery with a coil and developed a fatal rupture of the aneurysm ten days after proximal occlusion.

The patient was a 72-year-old woman who had complained of dysphagia and unsteadiness for several years. An approximately 20 mm diameter aneurysm was detected in her left vertebral artery. She underwent endovascular treatment, that is, her left vertebral artery was occluded with coils at a point proximal to the aneurysm. Her initial post-procedure course was uneventful. However, she suddenly developed right-side hemiparesis nine days after procedure. At that time, CT scan suggested sudden thrombosis of the aneurysm. Right vertebral angiography revealed a small part of the aneurysm. She was treated conservatively. Ten days after the procedure, she suffered massive subarachnoid haemorrhage.

Both the present case and past reports suggest that proximal occlusion of the vertebral artery is effective in treating relatively large aneurysms unsuitable for treatment by neck clipping or trapping. However, when the bifurcation of the posterior inferior cerebellar artery (PICA) is distal to the occluded point in cases where the PICA bifurcates from the aneurysm or the neck region, blood supply to the aneurysm may persist because anterograde blood flow to the PICA may be preserved. Therefore, clinicians must consider the possibility of aneurysm rupture after proximal occlusion in the following cases:

- 1) when the aneurysm is large or giant, but non-thrombosed;
- 2) when thrombosis occurs soon after the procedure;
- 3) when postoperative angiography shows partial filling of the aneurysm with contrast agent through the contralateral vertebral artery of basilar artery or the cervical muscle branches.

Introduction

Proximal occlusion of the vertebral artery is regarded as a safe and effective method of treating aneurysms of the vertebral artery or the vertebrobasilar junction unsuitable for treatment by neck clipping. In some cases, however, new neurological deficits can develop and pre-existing deficits can be exacerbated after this procedure.

These postoperative complications are often due to ischemia of the perforators or other re-

Figure 1 A) FLAIR axial MR image shows large aneurysm with a diameter of 20 mm with turbulent flow. B) T1-weighted sagittal image shows marked compression of brainstem, but no thrombus in the aneurysm.

gions. Very few reports have been published concerning early aneurysm rupture following proximal occlusion of the vertebral artery for cases of unruptured aneurysm. We recently encountered a case of large aneurysm of the vertebral artery diagnosed after initial detection of brainstem compression.

This patient underwent proximal occlusion of the vertebral artery with coils and suffered a fatal rupture of the aneurysm days after proximal occlusion.

Case Report

The patient was a 72-year-old woman who had occasionally experienced difficulty swallowing during meals for five years. She had also complained of dizziness while walking starting three years before. At that time, she consulted another clinic, where MRI revealed a mass compressing the brainstem. Angiography confirmed a diagnosis of left vertebral aneurysm. She was treated conservatively; however, her dysphagia progressed and the unsteadiness worsened. She was thus referred and admitted to our hospital.

Her consciousness was clear on admission. Although her motions were slightly slow, there was no evidence of motor disorder or pyramidal tract disturbance. She showed hoarseness, dysarthria and reduced pharyngeal reflex. Pha-

ryngeosophagography revealed disorders in the first to second phase of swallowing (in particular, severe disorder of the second phase). The maximum diameter of the mass, as measured by MRI and CT scans, was approximately 20 mm.

The mass severely compressed the anterior side of the brainstem (figure 1). Left vertebral angiography (figure 2) and three dimensional computed tomographic angiography (3D-CTA) revealed bifurcation of the posterior inferior cerebellar artery (PICA) from the base of the aneurysm and allowed us to determine that there was no thrombus within the aneurysm. Because the aneurysm was large, progressive, and appeared to be compressing the brainstem, we thought it necessary to take more aggressive measures against the aneurysm. The patient and her family declined craniotomy. We therefore selected endovascular treatment. Since the aneurysm was large and was compressing the brainstem, it seemed unlikely that intraaneurysmal embolization would arrest progression of symptoms.

The distance from the distal neck of the aneurysm to the union was only several millimeters, and it was difficult to accomplish endovascular trapping. Furthermore, if the treatment sacrificed the left PICA, her clinical symptoms would deteriorate. For these reasons, we elected to perform proximal occlusion of the left

Figure 2 Left vertebral angiography. Anteroposterior (A) lateral (B) and oblique (C) views show the PICA originating from the proximal neck of the aneurysm. The distance from the distal neck to union of the aneurysm was 2 to 3 mm.

vertebral artery with coils at a point immediately before the aneurysm, for the purpose of directly suppressing blood supply to the aneurysm.

Endovascular treatment was started under local anesthesia. First, the balloon occlusion test was performed for ten minutes. After observing no neurological changes and no change in the somatosensory evoked potential (SEP), the left vertebral artery was occluded with platinum coils at a point proximal to the aneurysm (figure 3). The woman developed no new neurological deficits after procedure. CT scans taken on the day following procedure showed a marked reduction in blood supply to the aneurysm as well as an indication of thrombosis of the aneurysm. From the day following procedure to four days after the procedure, heparin (10,000 units /day) was serially infused. Four days after the procedure, ticlopidine (200 mg/day) was started, and the heparin dose was gradually decreased.

Nine days after procedure, she suddenly developed hemiparesis of the right side. CT scans at that time suggested further thrombosis of the aneurysm (figure 4) and diffusion-weighted magnetic resonance images showed a strong signal in the brainstem.

These findings suggested brainstem ischemia due to damage of the perforators. Based on this judgment, the dose level of heparin was increa-

sed, and ozagrel sodium and edaravone were added to the drug therapy. On the following morning, her right side motor disorder was alleviated. However, right vertebral arteriography performed ten days after the procedure revealed partial filling of the aneurysm with contrast agent (figure 5). She continued to receive conservative treatment. In the early evening of the same day, she suddenly developed disturbance of consciousness.

At that time, CT scans revealed massive subarachnoid haemorrhage, possibly representing bleeding from the aneurysm (figure 6). She died 19 days after proximal occlusion of the vertebral artery.

Table 1 Cases of bleeding from large or giant vertebral artery aneurysm following proximal occlusion

	Age/ Sex	site of ligation	period between rupture and ligation
Yamada (1984)	40/F	proximal	16 months to PICA
Chang (1999)	72/F	proximal to aneurysm	9 days
Piepgas (2001)	27/F	C1-2 level	10 days
Iwabuchi (2004)	72/F	proximal to PICA	9 days

Figure 3 Immediate post-proximal occlusion angiographies show no filling of the left vertebral artery aneurysm from either the left vertebral artery (A) or contralaterally from the right vertebral artery (B).

Discussion

For many years, proximal occlusion of the vertebral artery has been used to treat aneurysms of the vertebral artery or the vertebro-

Figure 4 Noncontrast axial CT scan nine days after proximal occlusion shows extensive thrombus filling the aneurysmal sac.

basilar junction, which are difficult to treat by neck clipping^{1,2}. However, exacerbation of the condition (including onset of brainstem infarction and rerupture of aneurysm) sometimes occurs after proximal occlusion of this artery. Steinberg et al³ reviewed 201 patients who had undergone proximal occlusion for aneurysms of the vertebral artery or the basilar artery, which had been difficult to treat by clipping. They reported that the long-term prognosis was favorable in 73% of all cases and 86.5% of cases with vertebral aneurysms. In addition, they reported that the percentage of patients showing exacerbation of neurological deficits within four weeks after procedure was lower for patients in the vertebral aneurysm group (5.4%) than for patients with aneurysms of the basilar apex (32.5%), aneurysm of the basilar trunk (21.6%) or aneurysms of the vertebro-basilar junction (31.4%). On the basis of these results, Steinberg et al³ concluded that proximal occlusion was safer and more effective in treating cases of vertebral aneurysms than cases of aneurysms of the basilar artery or the vertebrobasilar junction. It is plausible to suggest that the outcomes of this therapy have further improved in recent years with the introduction of endovascular treatment, which can minimize postoperative complications.

The present case was initially followed without craniotomy since she was elderly and elected against craniotomy. However, since her symptoms worsened and the size of the aneurysm gradually increased, we decided to perform proximal occlusion of the parent artery using endovascular treatment.

Factors responsible for exacerbation of symptoms following proximal occlusion include ischemic complications and rupture of the aneurysm. However, bleeding after proximal occlusion was observed in only one (2.7%) of the 37 patients with vertebral aneurysms among the 201 cases reviewed by Steinberg et Al³. Furthermore, although several reports of rebleeding after proximal occlusion in cases of ruptured dissecting aneurysm of the vertebral artery have been published³⁻⁵, our literature search identified only four cases (including the present case) of saccular type aneurysm of the vertebral artery which suffered rupture after proximal occlusion.

Table 1 summarizes these previous reports. Yamada et Al⁶ analyzed the postoperative course of patients with vertebral aneurysms unsuitable for treatment by clipping with respect to the site where proximal occlusion of the vertebral artery was performed. They experienced one patient with giant vertebral aneurysm whose initial symptom was unsteady gait and who developed fatal subarachnoid haemorrhage 16 months after proximal occlusion of the vertebral artery. Yamada et Al⁶ reported that although the postoperative course was favorable in patients who underwent occlusion of the vertebral artery at a point distal to the PICA, brainstem infarction and subarachnoid haemorrhage were more likely to develop following occlusion of the vertebral artery at a point proximal to the PICA because blood supply from the vertebral artery to the PICA persisted and thrombosis of the aneurysm was not adequately induced. Chang et Al⁷ performed proximal occlusion of the right vertebral artery with a detachable balloon in a patient with giant vertebral aneurysm whose initial signs were headache, progressive ataxia and lower extremity weakness.

This patient developed disturbance of consciousness nine days after proximal occlusion and then developed fatal subarachnoid haemorrhage on the following day. In this case, the right PICA and anterior inferior cerebellar artery (AICA) originated from a common trunk

Figure 5 Right vertebral angiography ten days after proximal occlusion. Anteroposterior (A) and lateral (B) views show small retrograde filling of the aneurysm.

at the base of the basilar artery distal to the aneurysm. Although left vertebral angiography taken immediately after the procedure did not reveal the aneurysm, the same angiography nine days after the procedure showed that a small portion of the giant right vertebral aneurysmal base had recanalized and was filling from the opposite left vertebral artery injection. The right vertebral artery remained occluded. Chang et Al⁷ emphasized that even those patients with complete absence of aneurysm filling on angiographies immediately after proxi-

Figure 6 Noncontrast axial CT scan ten days after proximal occlusion shows massive subarachnoid haemorrhage.

mal occlusion should be followed up closely, particularly regions such as the basilar apex and distal vertebral artery where recanalization is more likely to occur from abundant collateral flow, and that such cases need to be followed by imaging until complete thrombosis of the aneurysm is confirmed. Piepgras et al⁸ also performed proximal occlusion of the right vertebral artery with a balloon in a case of giant aneurysm of the right vertebral artery whose initial symptoms were upper extremity weakness, incoordination, unsteadiness of gait and dysphagia. They reported that this patient developed left-sided hemiparesis and bradycardia and postural hypotension five days after the procedure and subarachnoid haemorrhage ten days after the procedure. In this case, the left vertebral artery did not contribute to the intracranial circulation, but widely patent posterior communicating arteries were noted. The right carotid artery arteriography, taken immediately after proximal occlusion, demonstrated retrograde flow along the basilar artery with faint retrograde opacification of the superior aspect of the aneurysm. MR angiography, performed before bleeding, also showed a residual channel of blood flow along the lateral and superior aspect of the aneurysm.

After bleeding developed, this patient underwent trapping under circulatory arrest. Sac disruption and thrombus extrusion from the aneurysm were visible intraoperatively. This patient experienced exacerbation of neurological deficits within 48 hours after discontinuation of heparin therapy. Piepgras et al⁸ interpreted this as evidence of aneurysm expansion due to rapid thrombus formation.

We analyzed these reported cases (including the present case) of unruptured vertebral aneurysm where clipping was difficult and bleeding developed after proximal occlusion of the vertebral artery.

These cases were found to have the following common features:

- 1) the aneurysm was large or giant but was non-thrombosed before the procedure;
- 2) the aneurysm became rapidly thrombosed after the procedure;
- 3) postoperative angiography showed partial filling of the aneurysm with contrast agent via the contralateral vertebral artery, the basilar artery or the cervical muscle branches.

Both the case reported by Piepgras et al⁸ and the present case showed exacerbation of neurological deficits after the heparin dose was reduced or discontinued. One possible means to deal with such a complication may be to extend anticoagulant therapy after procedure, to avoid rapid thrombosis of the aneurysm. However, additional study is needed to demonstrate this. The findings suggest that the PICA branching area should also be occluded when proximal occlusion is performed, after some reconstructive surgery has been taken for the PICA. However, whether this procedure can be safely performed depends on the features of individual cases.

Our experience with the present case and our review of past reports suggest that proximal occlusion of the vertebral artery remains a valid means of treating relatively large aneurysms unsuitable for treatment by neck clipping or trapping. However, since the PICA bifurcates from the aneurysm or the neck region, anterograde blood flow to the PICA may be preserved and blood supply to the aneurysm may persist in cases where bifurcation of the PICA is distal to the occluded point. Therefore, clinicians need to be aware of the risk of aneurysm rupture after proximal occlusion in cases presenting with the three above-mentioned features.

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EDITORIAL COMMENT

The above report is a major contribution to the literature and a very appropriate topic to be included in Interventional Neuroradiology. The authors have carefully gone through the literature and highlighted key features regarding this important problem.

I agree with the authors' formulation regarding the three common features that may predispose to delayed aneurysm rupture after proximal occlusion. In addition to these three features, the authors should highlight the location of the PICA in relationship to these aneurysms in the distal vertebral artery. At the same time the authors can emphasize the importance of an endovascular "trapping" procedure for those aneurysms that are separate from the PICA. That is, if the aneurysm is distal to the PICA then trapping of the aneurysm may be feasible with preservation of the PICA since it is proximal to the occlusion site. For those aneurysms that are clearly proximal to the PICA origin it may be possible to do an endovascular "trapping" procedure allowing the PICA to fill by retrograde flow. The problem, as highlighted in this case, is when the PICA arises at the neck of the aneurysm. In these cases a judgment must be made whether proximal occlusion is safe or whether a PICA-PICA by-pass should be done in conjunction with surgical or endovascular trapping of the aneurysm.

The topic of anticoagulation in deconstructive therapy is important. The authors state that anticoagulation may prevent thrombotic complications associated with proximal occlusion of these large or giant aneurysms. In this paper, the patient was treated with heparin for four days following the proximal occlusion. The authors state that it may be necessary to extend the anticoagulation therapy. Yet we are still missing a post-procedural anticoagulation strategy with specific recommendations regarding the timing of the medication.

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EDITORIAL COMMENT

This is a well described and interesting paper dealing with early rupture of a large aneurysm at the vertebrobasilar junction treated by proximal vessel occlusion with coils. The patient treated unfortunately had a fatal outcome due to the severe SAH. The authors conclude that large nonthrombosed aneurysms, that rapidly thrombose after arterial occlusion and that show a partial filling of the aneurysm via the contralateral vertebral artery or anastomotic channels at control angiography are at risk for rupture.

The authors do not insist sufficiently in our opinion on the pretherapeutic anatomic analysis that might anticipate potential complications that have been described in the literature. Sacrifice of the proximal portion of the PICA can be performed safely if there is a good haemodynamic balance with the AICA and SCA. The territories of the PICA can be evaluated thanks to the origin of the vessel itself: the lower the PICA the more cerebellar it is, the higher the PICA the more medullary it is. Some of the complications noted could have been avoided thanks to these observations. Furthermore, persisting flow in the aneurysm can be considered at short term FU angiographies as a negative sign: the redistribution of the flows after sacrifice, the persisting flow inside a small aneurysmal portion and the potential dissecting phenomena may contribute to the poor evolutions these patients have presented. Sacrifice of the parent vessel can be proposed safely and still belongs to the therapeutic panel to be proposed: it has to be performed in safe anatomical and technical conditions that might include (even loose) coiling of the sac itself with coils bulging into the parent vessel and occluding it. This might favour secondary safer aneurysm thrombosis and prevent rebleeds, especially if a dissecting phenomenon is suspected. The association of stents and coils might represent a better therapeutic alternative in selected cases, as would covered stents when they are made available for intracranial navigation.

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